

Brief Communications

Ruptured bronchial artery aneurysm associated with sarcoidosis

Hon Chi Suen, MD,^a Charles C. DuMontier, MD,^b John Boeren, MD,^c
Wayne Charland, MD,^d and Bill B. Daily, MD, PhD,^a Belleville, Ill



Dr Suen

Aneurysms of the bronchial arteries are rare, but their causes are diverse.¹ Bronchial artery aneurysms have never been described in association with sarcoidosis. We report the case of a 50-year-old woman with life-threatening rupture of a bronchial artery aneurysm associated with sarcoidosis.

Clinical Summary

A 50-year-old woman who underwent an uneventful coronary artery bypass 6 months previously had a 2-day history of back pain, nausea, and malaise. Then she experienced severe dizziness and was brought to the emergency department. She was found to be pale. Her vital signs included blood pressure of 188/88 mm Hg, heart rate of 105 beats/min, and respiratory rate of 28 breaths/min. Breath sounds were absent over the left side of the chest. There was no cardiac murmur. Peripheral pulses were all palpable. Chest radiography showed that the left side of the chest was completely opacified, with the mediastinum deviated to the right. The hemoglobin level was only 4.5 g/dL. Left-sided hemothorax was diagnosed, and placement of a tube in the left side of the chest yielded 1400 mL of frank blood. She had a history of renal artery stenosis, with a baseline serum creatinine level of 1.7 mg/dL. Acute renal failure developed, superimposed on chronic renal failure, with a creatinine level increasing to 3.1 mg/dL. As a result, the emergency physician ordered a computed tomographic (CT) chest scan without intravenous contrast material. It revealed a massive left-sided hemothorax and a large subcarinal mediastinal mass. She was resuscitated and given a transfusion. Chest tube drainage had slowed, and the creatinine level decreased to 2 mg/dL. A CT chest scan with intravenous contrast material showed no evidence of aortic aneurysm or dissection, but there was a small globular vascular structure below the carina with extravasation (Figure 1). The chest was then surgically explored.

Through a left thoracotomy through the fifth intercostal space, 2000 mL of blood clot was evacuated from the pleural cavity. A large mediastinal hematoma was identified between the heart and descending aorta, corresponding to the large subcarinal mass seen on the noncontrast CT scan. When the mediastinal hematoma was explored, many enlarged subcarinal and inferior pulmonary ligament lymph nodes were found, and pathologic examination revealed noncaseating granuloma consistent with sarcoidosis. Among the enlarged subcarinal lymph nodes, an actively bleeding 8-mm-diameter bronchial artery aneurysm was found. The feeding bronchial artery was ligated, and the aneurysm was excised. Pathologic examination confirmed a true aneurysm. Both staining and culture of the lymph nodes showed no evidence

From the Departments of Cardiothoracic Surgery,^a Radiology,^b Family Practice,^c and Pathology,^d St Elizabeth Hospital, Belleville, Ill.

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Address for reprints: Hon Chi Suen, MD, Cardiothoracic Surgery Associates, SC, 12B Park Place, Swansea, IL 62226 (E-mail address: HSUEN@earthlink.net).

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Figure 1. A CT chest scan with intravenous contrast showed no aortic pathology, but there was a bronchial artery aneurysm (arrow) below the carina with extravasation (arrowhead).

of fungal or acid-fast bacterial infection. She recovered uneventfully after surgical intervention. Mediastinal lymph node enlargement was her only manifestation of sarcoidosis. Repeat CT chest scanning 6 months later showed that mediastinal lymphadenopathy had resolved, and there was no other evidence of bronchial artery aneurysm.

Discussion

Bronchial artery aneurysm is a rare condition, with fewer than 30 cases reported in the literature. The aneurysms can be mediastinal or intraparenchymal in location. Mediastinal bronchial artery aneurysms can cause pain and hemothorax mimicking ruptured aortic aneurysm or dissection, superior vena caval obstruction caused by direct compression by a hematoma, or hematemesis when it ruptures into the esophagus. Intraparenchymal bronchial artery aneurysms usually present with hemoptysis. Ruptured bronchial artery aneurysm is a life-threatening condition. Bronchial artery aneurysms occasionally are asymptomatic and are detected as incidental findings on radiologic examination. A detailed review of this condition was provided by Kalangos and colleagues¹ in 1997.

Various causes of bronchial artery aneurysms have been cited. High bronchial arterial blood flow with resulting dilatation of the bronchial artery is the most frequently named pathogenetic process. Conditions related to this include bronchiectasis,² silicosis, and pulmonary artery agenesis.³ Infection by tuberculosis and syphilis has caused bronchial artery aneurysms. Bacterial infection and trauma have resulted in bronchial artery pseudoaneurysm formation and hemoptysis. Others were caused by atherosclerosis, Osler-Weber-Rendu syndrome, or medial degenerative changes,¹ but it could be idiopathic.⁴

Cardiovascular diseases have been described in association with sarcoidosis, but bronchial artery abnormality has never been reported. Sarcoidal angiitis has caused fibrosis in the adventitia of

the renal artery, resulting in stenosis. Aneurysms of the subclavian artery, abdominal aorta, or left ventricle have been reported in association with sarcoidosis. The multiple enlarged mediastinal lymph nodes with noncaseating granulomatous inflammation in our patient could have resulted in increased bronchial artery blood flow and a weakened arterial wall, resulting in aneurysm formation.

Bronchial artery aneurysms should be treated once they are diagnosed because such patients are at risk of life-threatening hemorrhage.¹ The size of the aneurysm has no bearing on its likelihood of rupture. Surgical excision of the aneurysm alone or segmentectomy, lobectomy, or pneumonectomy is the most secure way of extirpating the condition.⁵ Preoperative embolization has been reported to decrease intraoperative blood loss during elective operations.⁴ Embolization of the aneurysm alone without resection risks recurrence of the condition but is a good alternative in patients who are not surgical candidates.

References

1. Kalangos A, Khatchatourian G, Panos A, Faidutti B. Ruptured mediastinal bronchial artery aneurysm: a dilemma of diagnosis and therapeutic approach. *J Thorac Cardiovasc Surg.* 1997;114:853-6.
2. Yanagihara K, Ueno Y, Kobayashi T, Isobe J, Itoh M. Bronchial artery aneurysm. *Ann Thorac Surg.* 1999;67:854-5.
3. Sancho C, Dominguez J, Escalante E, Hernandez E, Cairois M, Martinez X. Embolization of an anomalous bronchial artery aneurysm in a patient with agenesis of the left pulmonary artery. *J Vasc Interv Radiol.* 1999;10:1122-6.
4. Nakajima H, Haneda T, Kambayashi M, Saitoh T, Yamada S, Takada N, et al. Two giant bronchial artery aneurysms: effect of preoperative embolization. *Eur J Surg.* 1995;161:855-6.
5. Saito Y, Ueda Y, Imamura H, Okamura A. Operative aneurysmectomy and middle lobectomy for asymptomatic bronchial artery aneurysm in young patient. *Eur J Cardiothorac Surg.* 2000;18:366-9.